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Case Report

Pathology Section

A Case Report of Two Malignancies in Breast with Short Review of Literature: Phyllodes and Ductal Type

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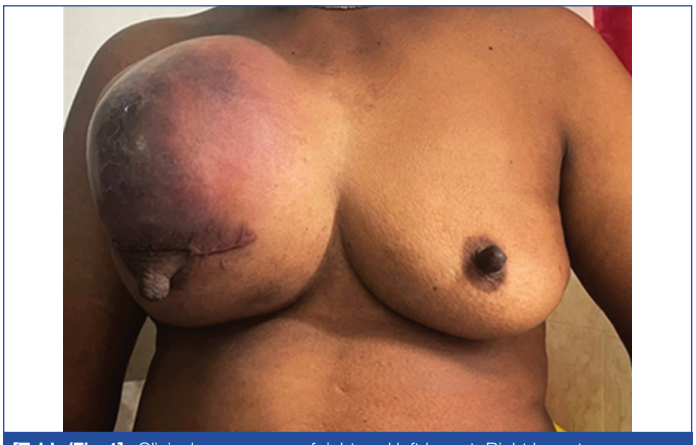
ABSTRACT

A 54-year-old female presented with a recurrent mass in the right breast for eight months, associated with pain and discolouration of the skin for two weeks. Past history revealed a wide local excision of a tumour in the same breast 10 months ago, which was reported as a benign phyllodes tumour. Local examination revealed a bosselated mass of size 20×20×10 cm occupying all four quadrants of the right breast, along with another firm lump of size 3×3 cm in the lower inner quadrant. Magnetic Resonance Imaging (MRI) of the breast showed two separate lumps: the large lump involved all four quadrants of the right breast and was suggestive of malignant transformation of a recurrent phyllodes tumour-BIRADS V (Breast Imaging-Reporting and Data System), and another smaller lump was noted in the lower inner quadrant. A Tru-cut biopsy of the larger lesion was reported as a possibility of a phyllodes tumour with mild nuclear atypia. The total mastectomy specimen showed both solid and cystic areas with necrotic material, haemorrhagic fluid, and blood clots in the larger lump. A separate lump from the lower inner quadrant of the ipsilateral breast showed a firm, solid lesion. Microscopic examination of both lesions revealed the simultaneous presentation of a malignant phyllodes tumour with chondrosarcomatous differentiation and infiltrating ductal carcinoma. This case highlights the unpredictable co-existence of two separate malignant lumps in the ipsilateral breast, which is an extremely rare event.

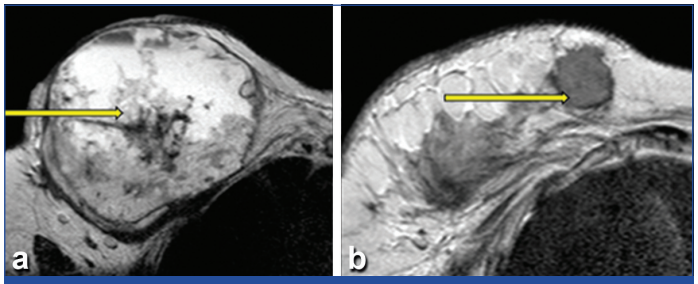
Keywords: Invasive ductal carcinoma, Phyllodes tumour, Two breast malignancies

CASE REPORT

A 54-year-old woman presented with a mass in the right breast for eight months and discolouration of the skin for two weeks. The lump was initially smaller in size but had a rapid increase in size over the past three months. A detailed history and physical examination revealed a past history of a wide local excision of the tumour in the same breast, which was reported as a benign phyllodes tumour. On examination, the right breast showed a bosselated mass measuring 20×20×10 cm occupying all four quadrants. A transverse hypertrophic healed scar measuring 12 cm was noted just superior to the areola in the same breast. Another firm lump measuring 3×3 cm was also noted in the lower inner quadrant of the same breast [Table/Fig-1]. There was no discharge from the nipple, and no lymph nodes were palpable in the right axilla. The left breast and axilla were normal. MRI of the right breast, plain and with contrast, revealed two separate lesions. The larger lesion appeared well-defined, solid-cystic with areas of haemorrhage, and showed chest wall invasion, suggestive of malignant transformation of recurrent phyllodes (BIRADS-V). The smaller separate lesion in the right breast was also suggestive of BIRADS-V [Table/Fig-2]. A Trucut biopsy of the larger lesion revealed the possibility of a phyllodes tumour with mild nuclear atypia.

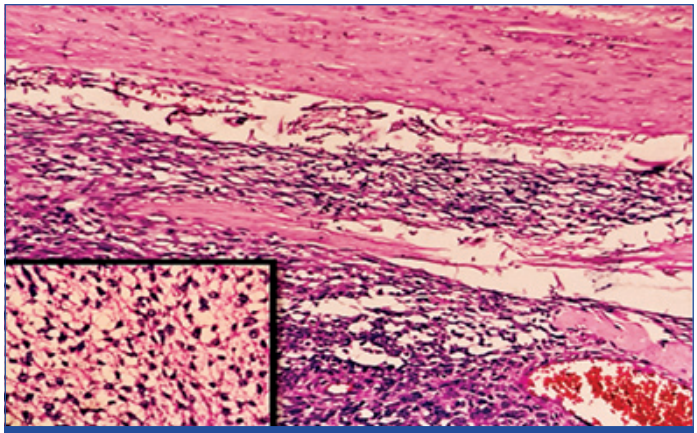


[Table/Fig-1]: Clinical appearance of right and left breast. Right breast appears – bosselated, erythematous with a large breast mass displacing the nipple areola complex inferiorly. Healed scar is located just above the areola.

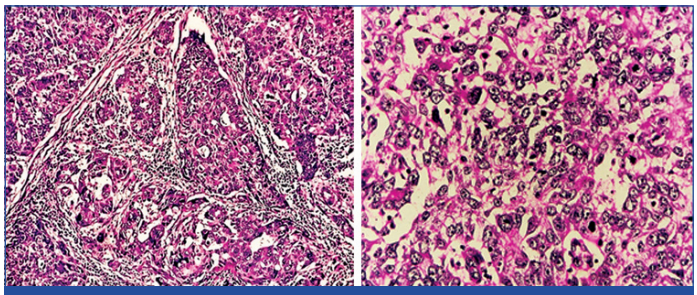


[Table/Fig-2]: MRI examination: a) Larger tumour: T2 hyperintense lesion in right breast exhibiting cystic component; b) Smaller tumour: T2 hyperintense lesion in the lower inner quadrant of right breast.

Total mastectomy was performed. Two separate specimens and three axillary lymph nodes were received. In the larger specimen, grossly, the skin surface showed a bulge and brownish discolouration in the central zone, with nodulocystic consistency. The cut surface showed predominantly a cystic tumour with a peripheral solid rim measuring 12×10×7.5 cm. The cystic space contained necrotic material, haemorrhagic fluid, and blood clots. Another separate smaller firm mass measured 2.9×2.5×2 cm, and the cut surface showed a grey-white, solid, circumscribed tumour with an irregular outline. Microscopic examination of the larger cystic mass showed a tumour predominantly composed of a proliferating stromal component with capsular invasion and a heterologous malignant Grade-II Chondrosarcomatous component [Table/Fig-3]. The deep surgical margin was involved by the tumour. The smaller firm mass from the lower inner quadrant showed Grade-III poorly differentiated infiltrating ductal carcinoma [Table/Fig-4,5]. Microscopic examination of the 3 lymph nodes from the axilla showed no tumour deposits. Within the IDC, no lymphovascular invasion was noted. Following surgical management, the patient was referred to a higher centre for further management and was lost to follow-up. Thus, both types of malignant phyllodes and ductal carcinoma were found simultaneously.



[Table/Fig-3]: Malignant phyllodes – Proliferating stromal component showing invasion (H&E stain, X100). Inset showing chondrosarcomatous component (H&E stain, X400).



[Table/Fig-4]: Invasive ductal carcinoma from smaller mass (H&E stain, X100).
[Table/Fig-5]: Invasive ductal carcinoma from smaller mass (H&E stain, X400).
(Images from left to right)

DISCUSSION

Phyllodes tumours are categorised by factors such as stromal cellularity, stromal atypia, stromal overgrowth, mitosis, and tumour margins, resulting in benign, borderline, or malignant classification [1]. Malignant phyllodes tumours may exhibit a heterologous sarcomatous component. Additionally, any phyllodes tumour can potentially contain ductal carcinoma in situ or invasive carcinoma, although this occurrence is uncommon [2]. Present case reported the right breast tumour as malignant phyllodes based on invasive margins, high cellularity, composed of plump to elongated spindle-shaped cells arranged in fascicles and storiform pattern, moderate nuclear pleomorphism with a few multinucleated cells, high mitotic count (5/10 HPF), and a heterologous malignant mesenchymal Grade-II chondrosarcomatous component.

A total of 14 cases of two breast malignancies have been reported in English literature in the last 20 years to the best of authors knowledge. A summary of all cases and the present case is highlighted in [Table/Fig-6] [3-14]. Out of the 15 cases, including the present case, 10 cases showed ipsilateral occurrence, while the remaining five were contralateral. Co-existing carcinoma in malignant phyllodes tumour is more commonly ductal (invasive ductal carcinoma, n=14) than lobular (invasive lobular carcinoma, n=1). Out of the 10 ipsilateral cases, four cases of breast carcinoma were present within the malignant phyllodes, and six were present separately. Heterologous sarcomatous components were noted in four cases, and the present case showed a chondrosarcomatous component. Most of the cases showed metastasis to axillary lymph nodes.

Author/Publication year	Age (years)	Size of MPT (cm)	Location of MPT	Type of breast CA	Size of CA	Location of CA	Heterologous component	Metastasis	Outcome
Merck B et al., 2006 [4]	46	5	Right breast	Infiltrating ductal carcinoma	3.5 cm	Contralateral	NA	Left axillary lymphatic ganglions	Free of disease 32 months after surgery
Korula A et al., 2008 [5]	51	16	Left breast	Invasive ductal carcinoma + DCIS	NA	Ipsilateral (within)	NA	Left axillary lymph node	Free of disease – 11 months after surgery
Kefeli M et al., 2008 [6]	26	4.5	Left breast	Invasive ductal carcinoma	2.5 cm	Ipsilateral (separate)	Chondrosarcoma and Liposarcoma differentiation	Left axillary lymph node	Died after one year of diagnosis
Abdul Aziz M et al., 2010 [7]	43	3.5	Left breast	Invasive ductal carcinoma + DCIS, LCIS	2 mm	Ipsilateral (IDC- separate) (DCIS, LCIS-Within)	Liposarcomatous	NA	No metastasis after one year follow-up
Shin YD et al., 2014 [8]	45	24	Left breast	Invasive ductal carcinoma of NST	NA	Ipsilateral (Separate)	NA	Left axillary lymph node	Recurred after six months
Sato T et al., 2016 [9]	47	5.5	Left breast	Invasive ductal carcinoma	13 cm	Contralateral	NA	Left axillary lymph node, Lungs and liver	Died nine months after surgery
Muthusamy RK and Mehta SS, 2017 [10]	51	15.5	Left breast	Invasive ductal carcinoma of NST	2.5 cm	Ipsilateral (separate)	NA	Left axillary lymph node	On chemo and radiotherapy
To H et al., 2018 [11]	48	6.5	Right breast	Invasive lobular carcinoma	1.5 cm	Ipsilateral (Separate)	NA	Right axillary lymph node	Recovered well and on follow-up
Nistor-Ciurba CC, 2020 [12]	50	11	Left breast	Invasive ductal carcinoma + DCIS	NA	Ipsilateral (within)	Chondrosarcomatous	NA	Follow-up for 11 years with no recurrence
	71	5	Right breast	Invasive ductal carcinoma of NST	NA	Ipsilateral (within)	NA	NA	Alive for 3 years + 3 months. Lost follow-up
	75	4	Left breast	Invasive ductal carcinoma of NST + DCIS	2 cm	Ipsilateral (within)	NA	NA	Follow-up three years. Died after 4 and ½ years of initial treatment.
Nath AR et al., 2021 [3]	81	4	Left breast	Invasive ductal carcinoma of NST	1.5 cm	Contralateral	NA	NA	Under follow-up
Sinuraya LW and Yarso KY, 2022 [13]	45	6.5	Right breast	Infiltrating ductal carcinoma, NST	2.8 cm	Contralateral	NA	NA	Under follow-up

Ray M et al., 2023 [14]	64	15	Left breast	Invasive ductal carcinoma + DCIS	5 cm	Contralateral	NA	Right axillary lymph node, Left lung and brain	Referred to palliative care
Present case (2024)	54	12	Right breast	Invasive ductal carcinoma – NST	2.9 cm	Ipsilateral (separate)	Chondrosarcomatous	NA	Referred to higher centre for chemotheraphy

[Table/Fig-6]: Summary of reported cases of two malignant tumours in breast: MPT and Carcinoma breast.
MPT: Malignant Phyllodes tumour; IDC: Invasive ductal carcinoma; NST: Non specific type; DCIS: Ductal carcinoma Insitu; LCIS: Lobular carcinoma Insitu; NA: Not available

The molecular events associated with the transformation and progression of phyllodes tumours remain largely unknown. The infrequency of co-existing epithelial malignancy complicates the ability to make definitive conclusions regarding the aetiological association between carcinoma and phyllodes tumours in these cases [7]. Malignant transformation of epithelium results from stromal-epithelial interactions within the phyllodes tumours or cancerisation of a phyllodes tumours by carcinoma arising in the duct system peripheral to the phyllodes tumours. This explains carcinoma present within the phyllodes tumours as well as peripheral to it in the ipsilateral breast [7]. The separate tumour in the ipsilateral breast or tumour arising in the contralateral breast may indicate a genetic predisposition for double primary breast cancer. This tendency could be linked to inherited mutations in genes like BRCA1/2 and potentially other genetic alterations [13]. In the present case, IDC was found separate from the malignant phyllodes tumours. However, the patient had a benign phyllodes tumour that had recurred. Hence, there is a possibility that epithelial malignancy might be a part of the remnant of the initial phyllodes. The clearance from the surgical margin was only 1 mm in the initial resection for benign phyllodes tumours.

The mainstay of treatment for phyllodes tumours includes wide local excision with a minimum margin of 1 cm or mastectomy. Clear margins are crucial for preventing recurrence, especially in borderline and malignant phyllodes tumours. Adjuvant chemotherapy and radiotherapy are considered for malignant cases, with individualised approaches for co-existing breast carcinomas [15].

CONCLUSION(S)

As the entity is rare, two malignancies are possible in the ipsilateral or contralateral breast. Also, the surgical treatment of phyllodes is wide local excision. If specimen is received with a margin <1 cm, the histopathology report should highlight this and discuss it with the surgeon.

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